

Limb Salvage of a Flail Neuropathic Ankle in Congenital Insensitivity to Pain with Anhidrosis Using a Hindfoot Superconstruct

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Learning Point of the Article:

Hindfoot arthrodesis using the superconstruct principle can successfully salvage a neuropathic flail ankle secondary to congenital insensitivity to pain with anhidrosis and restore independent ambulation.

Abstract

Introduction: Congenital insensitivity to pain with anhidrosis (CIPA) is a rare autosomal recessive neurological disorder, with an estimated incidence of 1 in 125 million. The condition is characterized by a clinical feature of unexplained pyrexia, global insensitivity to pain, anhidrosis, intellectual disability, and self-mutilating behavior. While musculoskeletal complications, such as fractures and joint dislocations are common, severe neuropathic arthropathy represents one of the most debilitating skeletal manifestations; however, its specific incidence remains unrecorded in present medical literature. Due to progressive joint destruction and gross instability, limb salvage and functional restoration remain a significant surgical challenge.

Case Report: A tertiary care orthopedic center reported the case of a 24-year-old female, who was wheelchair-bound for 7 years and presented with a painless, severely deformed, and unstable ankle complicated by a history of recurrent infections. A diagnosis of a flail neuropathic ankle secondary to CIPA was established. To achieve limb salvage, the patient underwent hindfoot arthrodesis utilizing the “superconstruct” principle. This approach aimed to provide a stable, plantigrade foot through rigid internal fixation and intensive post-operative rehabilitation. Clinical and radiological union was well-established by 10 weeks, at which point assisted mobilization was achieved. At the 1-year follow-up, radiological assessment confirmed solid bony fusion, and the patient was successfully ambulating without assistance.

Conclusion: Managing a neuropathic flail ankle in the context of CIPA requires meticulous pre-operative planning, a detailed diagnostic workup, and aggressive post-operative management with respect to wound care and rehabilitation. This case demonstrates that hindfoot arthrodesis utilizing a plate-and-screw superconstruct provides the mechanical stability necessary to overcome the biomechanical challenges of CIPA, proving that limb salvage and functional restoration are achievable goals.

Keywords: Congenital insensitivity to pain, neuropathic ankle, charcot arthropathy, superconstruct, arthrodesis, limb salvage.

Introduction

Congenital insensitivity to pain with anhidrosis (CIPA) is a rare genetic disorder—with an estimated incidence of 1 in 125 million—defined by a complete absence of nociception alongside a systemic inability to sweat. This sensory deficit frequently results in repetitive microtrauma, culminating in

progressive joint destruction and complex deformities secondary to untreated and neglected fractures [1,2,3]. They often present with multiple musculoskeletal deformities following fractures and dislocations. Neurological disorders of this nature often manifest in the lower extremities as severe instability and profound functional impairment. From the

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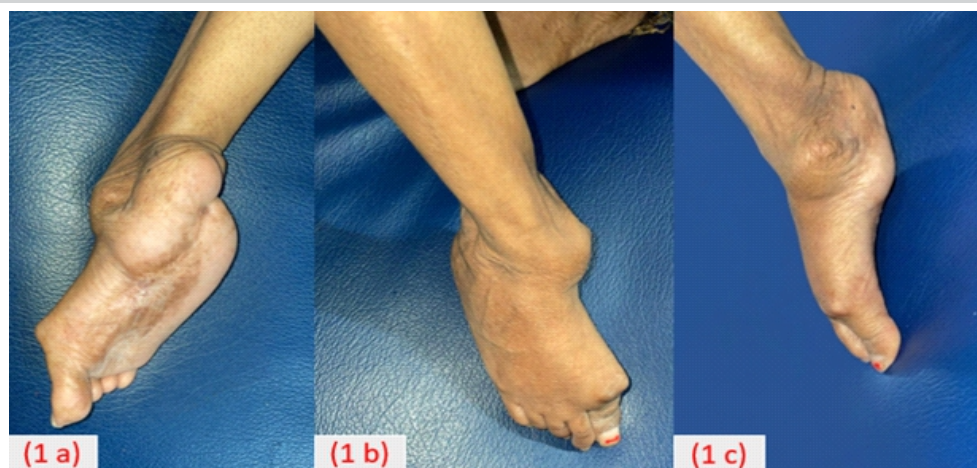


Figure 1: Clinical image of the patient's foot: Plantar (a), Dorsum (b), and Lateral (c) surfaces of the right foot.

surgeon's perspective, managing these cases is exceptionally demanding due to the combination of distorted anatomy, poor bone mineral density, accelerated bone resorption, and gross instability [4,5]. While hindfoot arthrodesis is the present established gold standard treatment for irreducible or non-braceable neuropathic deformities, standard implants often result in construct failure because of the underlying biological and mechanical compromises [6,7]. This report outlines the successful management of a uniquely complex neuropathic ankle using an extended fusion "superconstruct" strategy—a technique that, to our knowledge, is yet to be documented in the medical literature.

Case Report

The patient, a 24-year-old female who had been wheelchair-



Figure 2: Radiograph of ankle joint anteroposterior view (a) and lateral view (b).

bound for 7 years, presented to a tertiary care orthopedic center, with a grossly deformed and highly unstable flail ankle (Fig. 1). Her clinical history was significant for recurrent localized infections and a series of multiple fractures in the past that resulted in deformities of the upper limbs. Upon physical examination, she demonstrated a complete absence of pain during joint manipulation, a hallmark finding that led to a detailed clinical evaluation and the subsequent diagnosis of CIPA. On examination, the patient exhibited severely dry skin and a Tendo-

Achilles contracture alongside an unstable flail ankle and flexion contractures of the toes. The combination of a biologically compromised limb and a mechanically unstable joint presented a unique therapeutic hurdle to the treating orthopedic surgeon.

Radiographic evaluation revealed severe regional osteopenia, grossly distorted ankle anatomy with an atrophic boat-shaped calcaneum, and complete resorption of the talus, consistent with advanced Charcot neuroarthropathy (Fig. 2). Computed tomography with 3D reconstruction (Fig. 3) and 3D printing was done to aid in pre-operative planning.

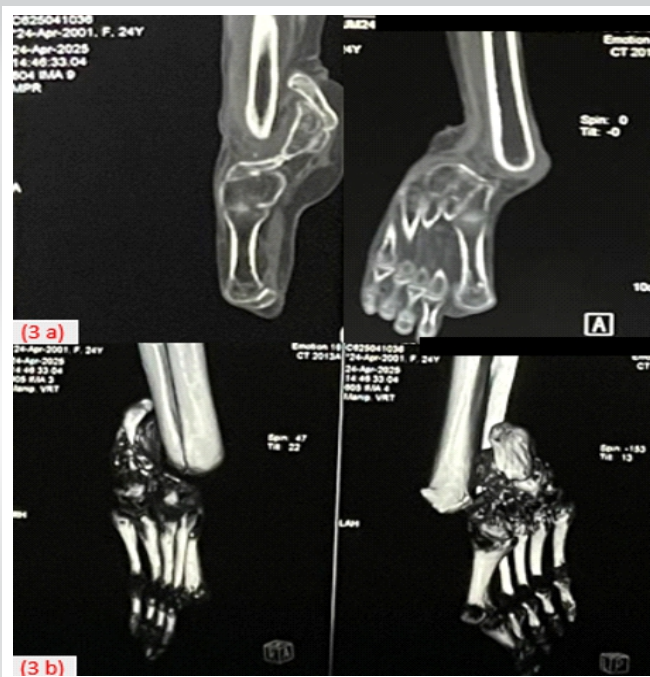


Figure 3: Computer tomography (a) images and 3D reconstruction (b).



Figure 4: Surgical step showing transfibular approach after distal fibular osteotomy.

The primary surgical objective was limb salvage through stable hindfoot arthrodesis, aimed at achieving a plantigrade foot capable of functional weight-bearing. The surgical challenges were multifaceted: A profound neuropathic state predisposing to delayed union; compromised bone stock increasing the risk of hardware pull-out and construct failure; severely distorted



Figure 6: C-arm images showing tibio-calcaneal arthrodesis using beam screws.



Figure 5: Preparation of tibio-calcaneal arthrodesis site.

anatomy; a compromised soft-tissue envelope; and a significant Tendo-Achilles contracture necessitating a formal release.

Pre-operative planning focused on the surgical approach, implant selection, and optimal hardware positioning. Several internal fixation modalities were evaluated during the planning phase. Tibio-talo-calcaneal intramedullary nailing was precluded by an exceptionally narrow medullary canal and a severely deformed calcaneus, compounded by the complete absence of the talus. Consequently, an open tibio-calcaneal arthrodesis was performed using a modified Mann's technique. Stability was ensured through a rigid "superconstruct" comprising beam screws and a fusion plate, providing the necessary mechanical environment to facilitate fusion in a neuropathic limb.

Surgical intervention commenced with a Hoke triple hemisection of the Achilles tendon to correct the equinus



Figure 7: Intraoperative picture after anterior fusion plate fixation and repositioned lateral malleolus.



Figure 8: Post-operative radiograph showing anteroposterior (a) and lateral (b) views.

contracture. A lateral transfibular approach was employed, as described in the modified Mann's technique, utilizing a fibular osteotomy performed approximately 5 cm proximal to the ankle joint. After osteotomizing its medial one-third (Fig. 4) to freshen the surface, the distal fibular segment was reflected posteriorly, with careful attention paid to preserving its vascular supply for subsequent use as an onlay graft. This provided direct visualization and allowed for meticulous preparation of the tibiocalcaneal arthrodesis site (Fig. 5). To promote osteogenesis, autologous corticocancellous bone graft was harvested from the ipsilateral iliac crest and packed into the fusion site. Initial stabilization and compression were achieved using headless, variable-pitch compression screws directed cephalad from the calcaneus into the tibia (Fig. 6).

The "superconstruct" principle was implemented through extended fusion, aggressive bone resection, and the use of high-strength implants in optimal alignment. This screw fixation was augmented with an anterior ankle fusion plate to establish a rigid, load-sharing construct. Furthermore, the reflected distal fibula was utilized as a vascularized onlay graft bridging the lateral cortices of the tibia and calcaneus, secured with two cortical screws (Fig. 7). Concurrently, a symptomatic hallux flexus deformity was addressed through a flexor hallucis longus tenotomy and K-

wire arthrodesis of the first metatarsophalangeal joint. Post-operative radiography confirmed optimal implant positioning and the successful restoration of plantigrade alignment (Fig. 8).

On post-operative day 5, the patient developed marginal skin necrosis. This necessitated surgical debridement, during which one exposed screw was removed. The wound was managed successfully with a vacuum-assisted closure (VAC) device and subsequently reconstructed with a split-thickness skin graft (STSG) (Fig. 9).

Post-operatively, the limb was immobilized in a below-knee cast for 3 months. A strict weight-bearing protocol was enforced: Absolute non-weight-bearing for the initial 6 weeks, progressing to toe-touch weight-bearing, and gradually advancing to full weight-bearing by 3 months. A custom

orthosis was prescribed for long-term wear to protect against future neuropathic breakdown and limb length variation of 1.5 cm. At the 3-, 6-, and 12-month follow-ups, solid bony consolidation was evident both clinically and radiographically (Fig. 10). After 7 years of wheelchair dependency, the patient successfully regained the ability to ambulate independently without assistance. American Orthopaedic Foot and Ankle Society improved from 30 pre-operatively to 81 at 1 year follow-up.

Discussion

CIPA is an extremely rare genetic entity [1,4], characterized by a systemic lack of nociception frequently manifesting as a series



Figure 9: Post-operative skin necrosis on day 5 (a), which subsequently underwent debridement and Vacuum assisted closure (b) and Split Thickness skin grafting (c).



Figure 10: 1-year follow-up showing anteroposterior (a) and lateral view (b) radiographs with clinical image (c) and patient ambulation (d).

of unrecognized musculoskeletal injuries, including recurrent fractures and dislocations that inevitably progress to severe deformities and neuropathic arthropathy. On review of the literature, clinical reports of this condition remain sparse; consequently, the management of the resulting joint destruction is often highly empiric, relying heavily on localized institutional experience rather than standardized protocols [2]. The absolute absence of protective sensation leads to repeated microtrauma, unchecked joint destruction, and classic Charcot-like neuroarthropathy. In advanced stages, this presents as a grossly deformed, non-braceable, and flail limb necessitating amputation [3,5].

Traditional treatment methods for patients with severe bony resorption, compromised soft tissue envelopes, and gross instability often made amputation the most predictable – and sometimes the only – viable treatment option for the resultant neuropathic joint [7]. While hindfoot arthrodesis is the gold standard for limb salvage, traditional fusion techniques in neuropathic conditions are fraught with unacceptably high complication rates, including non-union, implant failure, and surgical site infection [6,8]. Indeed, non-union rates have been reported as high as 41% in the literature [9]. In patients with CIPA, the native bone quality is frequently characterized by profound osteopenia. This makes standard isolated screw fixation or intramedullary nailing insufficient to withstand the unopposed, abnormal mechanical stresses generated during weight-bearing. The challenge is further exacerbated by the patient's underlying biological compromise, which significantly impairs the healing response.

To counter this compromised biomechanical environment, we applied the “superconstruct” principles popularized by Sammarco [10]. This strategy mandates extending the fusion

beyond the immediate zone of injury, performing aggressive bone resection down to healthy, bleeding bone to correct the deformity, and utilizing the strongest possible hardware. In our case, the combination of headless compression screws for dynamic interfragmentary compression, augmented by a dedicated rigid fusion plate, created a robust load-sharing construct. This construct effectively neutralized deforming forces while maximizing the surface area for bony consolidation.

Furthermore, our choice of surgical approach provided a dual mechanical and biological advantage. We utilized a lateral transfibular approach employing a modified Mann's technique [11]. This not only offers extensive, direct visualization for thorough joint debridement and deformity correction but also yields the resected distal fibula for use as an onlay vascularized fibular graft. In neuropathic joints where the biologic healing cascade is often severely blunted, supplementing a rigid mechanical superconstruct with a robust local bone graft is a critical step in achieving the solid bony union observed at our patient's 1-year follow-up.

Finally, the surgical management of CIPA-associated neuropathic ankles extends well beyond osseous fixation; it is equally a soft-tissue challenge. The soft tissue envelope in these patients is notoriously fragile, and the patient's lack of pain eliminates the primary early warning sign of wound ischemia or impending breakdown. Our patient's complication on post-operative day 5 underscores this vulnerability. The successful salvage of the limb and the underlying hardware was entirely dependent on prompt recognition, aggressive surgical debridement, and the integration of VAC and a STSG. This highlights that achieving a union in these complex deformities requires a vigilant, multidisciplinary post-operative protocol.

Conclusion

Neuropathic ankles secondary to CIPA are exceedingly rare and present formidable reconstructive challenges. Nevertheless, this case demonstrates that limb salvage is achievable through hindfoot arthrodesis using a modified Mann's technique. By integrating compression screws with a plate superconstruct, surgeons can achieve the rigid stability necessary for successful fusion. Such an approach underscores that amputation is not an inevitability; rather, through robust reconstruction, it is possible to restore ambulation and dramatically improve functional outcomes. In the management of complex

neuropathic deformities, amputation should be the final chapter, not the first option.

Clinical Message

The primary clinical message is that limb salvage and functional restoration remain achievable, even in severe flail neuropathic ankles. This case demonstrates that hindfoot arthrodesis using a "superconstruct" can overcome challenges in congenital insensitivity to pain with anhidrosis, yielding excellent outcomes and restoring independent ambulation after years of wheelchair dependency.

Declaration of patient consent: The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given the consent for his/ her images and other clinical information to be reported in the journal. The patient understands that his/ her names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Conflict of interest: Nil **Source of support:** None

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